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RENAL HEMANGIOMA

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Four cases of renal hemangioma are presented. Renal hemangioma is difficult to detect because this benign vascular tumor never demonstrates any abnormalities on renal arteriography. Computed tomography in two resected cases of cavernous hemangioma revealed a low-density mass without any enhance effect, while the others diagnosed by the selective renal arteriography demonstrated no abnormality on computed tomography. We postulate that both angiographic and computed tomographic appearances of the renal hemangioma could depend on its vascular components. Related reports are also reviewed.

Key words: Hemangioma, Renal pelvic tumor, Renal pelvis, Kidney

INTRODUCTION

Hemangioma of the kidney is an uncommon and benign vascular tumor. Virchow was probably the first to document an autopsy case in 1867, and Fukuda described the first Japanese case from an autopsy in 1967[1]. To date a total of 50 cases has been found in the Japanese literature[2] But it is thought that the lesion is not as uncommon as the limited number of case reports suggests, and that renal hemangioma undoubtedly accounts for a certain percentage of those instances of renal bleeding diagnosed as "essential hematuria"[3]

Recently, we encountered 4 cases of renal hemangioma. Herein we report these cases with a review of the literature.

CASE REPORT

Case 1. A 64-year-old man visited our hospital, complaining of painless gross hematuria which had its sudden onset 3 days ago. He had no past episode of bloody urine. Cystoscopy revealed that the gross hematuria emanated from the right ureteral orifice. An excretory urography demonstrated deformity in the lower calices of the right kidney. This finding was confirmed by retrograde pyelography (Fig. 1). An abdominal computed tomography showed an abnormal mass measuring 2.0×1.5 cm in the right renal pelvis, which was not enhanced with contrast medium (Fig. 2). Aortography revealed three arteries of the right kidney, and these vessels had neither obstruction, hypervascularity nor encase-

ment. ^{67}Ga-scintigraphy showed no abnormal accumulation. Repeated cytologic examinations of voided or ureteral urine were negative for malignant cells. The preoperative diagnosis was a right renal pelvic tumor, probably with benign nature. Right renal nephroureterectomy with bladder cuff removal was performed. On cut section there was a soft round tumor, 20 × 14 mm in diameter, partly filled with blood, in the lower calices (Fig. 3). Other renal tissue was, however, of normal appearance. Microscopically, tumor had multiple vascular spaces in the subepithelial region of lower calices (Fig. 4). These spaces were lined by flat endothelial cells with thin connective tissue bands. Pathological diagnosis was a cavernous hemangioma of the right renal pelvis. The patient is healthy and asymptomatic 2 years later.

Case 2. A 62-year-old man complained of gross hematuria and left lower abdominal discomfort prior to consultation. His medical history included a left pyeloplasty for the stricture of left uretero-pelvic junction 6 years previously. Cystoscopic examination revealed no abnormality in the vesical mucosa. An excretory urography showed dilated pelviocaliceal system of the left kidney. A retrograde pyelography with smooth catheterization demonstrated a filling defect in the left renal pelvis. Left renal sonography showed an echogenic mass pattern near the ureteropelvic junction in an opened central echo complex. Cytologic study of a sample taken from the left renal pelvis suggested class IIIb. Repeated cytologic evaluations of voided urine also demonstrated class IIIb or IIIa. The mass in the renal pelvis was unremarkable upon computed tomography, while a small cystic lesion was observed in the left medullary area (Fig. 5). RI-angiography revealed no pooling shadow. Despite medical treatment, bloody urine continued. A left nephroureterectomy was performed and a cuff of bladder was resected. The surgical specimen weighed 256 gm. Much coagula was obtained from the left renal pelvic cavity, and its mucosa showed severely hemorrhagic appearance on cut section (Fig. 6). There were macroscopically no tumorous lesions except for a few mucosal folds in the renal pelvis. Microscopically, there were foci of severe dysplasia made up of atypical transitional epithelia. A cavernous hemangioma which was depicted as a small cystic lesion upon computed tomography, was also disclosed upon further observation of additional sections (Fig. 7). Moreover, the so-called renal adenoma with *in situ* changes was encountered in the proximal convoluted tubule. Unfortunately, about 2 months after surgery, he died of multiple organ failure, mainly hepatic and renal failure in spite of intensive care.

Case 3. A 34-year-old woman had acute attack of right back pain and gross hematuria a day earlier. She also experienced the same episode 4 years ago, when hematuria diminished after several days with medical treatment. An IVP showed no excretion of contrast medium from the right kidney. The following retrograde pyelography confirmed filling defect considered as blood clots in the right renal pelvis. Urine cytologic studies were negative for malignant cells. Cross sectional images of computed tomography and sonography demonstrated no evident abnormality. Although a digital subtraction angiography revealed almost normal distribution of the right renal arteries (Fig. 8), right renal arteriography disclosed a coil of blood vessels at the arterial phase without an early venous drainage (Fig. 9). This shadow appeared 1 minute after injection of contrast medium, but pooling was unremarkable at the capillary or venous phase. We diagnosed it as a hemangioma of the right kidney. Hematuria disappeared 3 days after admission and then she was discharged from hospital. Follow-up examinations have been unchanged for one year.

Case 4. A 33-year-old woman was hospitalized with right back colic and gross hematuria, which had suddenly appeared several hours before admission. Blood coagulum and hemorrhage were endoscopically lateralized to the right side. The right kidney did not excrete the contrast medium upon IVP. The hematuria

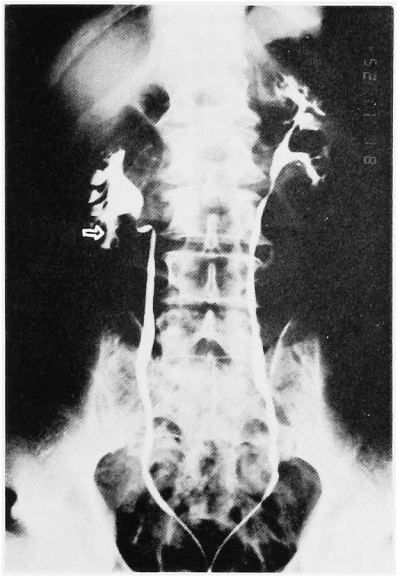

Fig. 1. Case 1—Retrograde pyelography shows a deformity with faint filling of contrast medium in the lower calices of the right kidney (arrow).

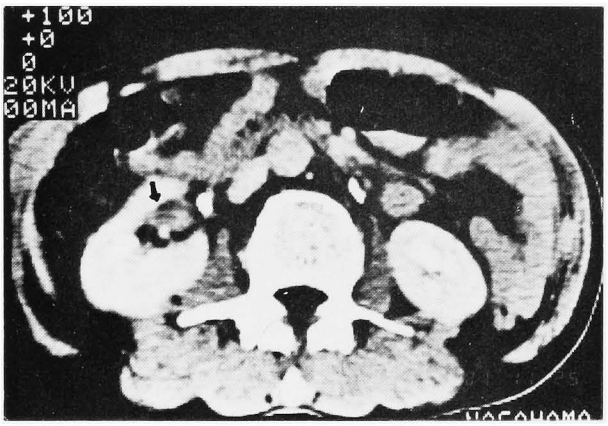

Fig. 2. Case 1—Computed tomography demonstrates a well-defined mass in the right renal pelvis (arrow). The mass was not enhanced with contrast medium.

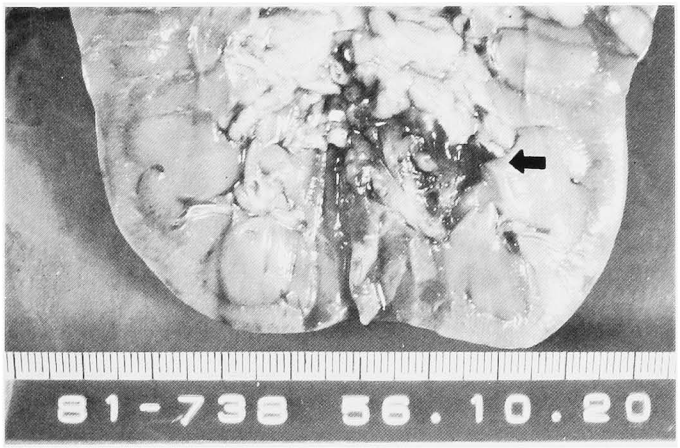

Fig. 3. Case 1—Macroscopic appearance of the cut specimen. There was a subepithelial tumor partly filled with blood in the lower calices (arrow).

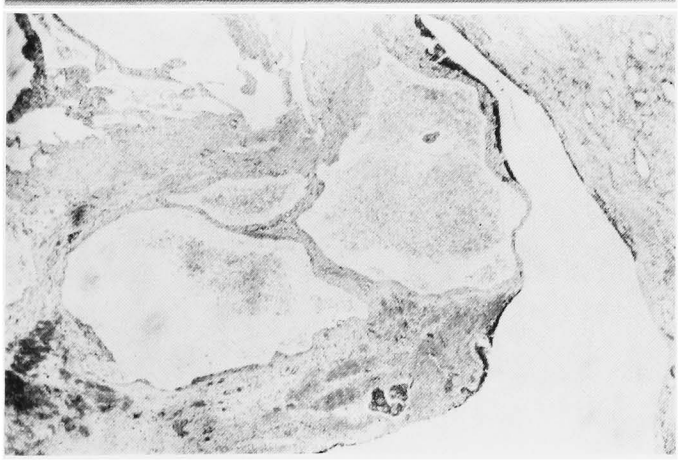

Fig. 4. Case 1—Microscopic appearance. The tumor was composed of vascular channels, characteristic of a cavernous hemangioma, in the subepithelial region. H.E. stain, ×40.

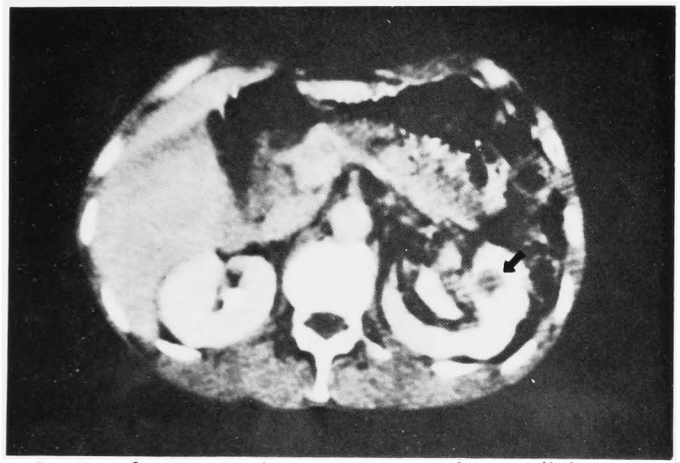

Fig. 5. Case 2—Computed tomogram reveals a well-demarcated, spherical and low-density mass in the medullary area of the left enlarged kidney (arrow). This cystic region was retrospectively regarded as the position where a cavernous hemangioma was encountered.

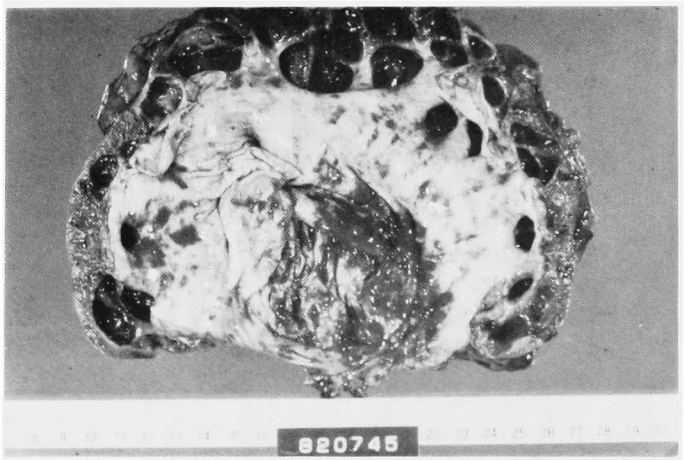

Fig. 6. Case 2—Macroscopic appearance. Subepithelial hemorrhage seen in the left renal pelvis, but there appear to be no tumorous lesions except for a few mucosal folds.

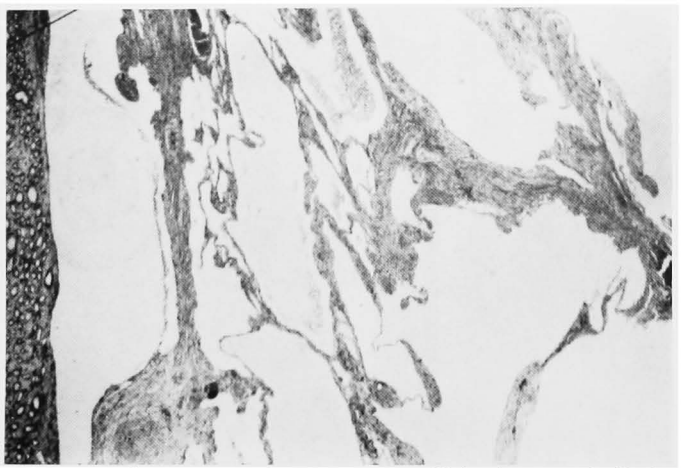

Fig. 7. Case 2—Microscopic appearance. This photograph discloses a cavernous hemangioma adjacent to the normal papilla. H. E. stain, ×40.

Fig. 8. Case 3—Digital subtaction angiography demonstrates no evident abnormality such as blush, distortion or pooling

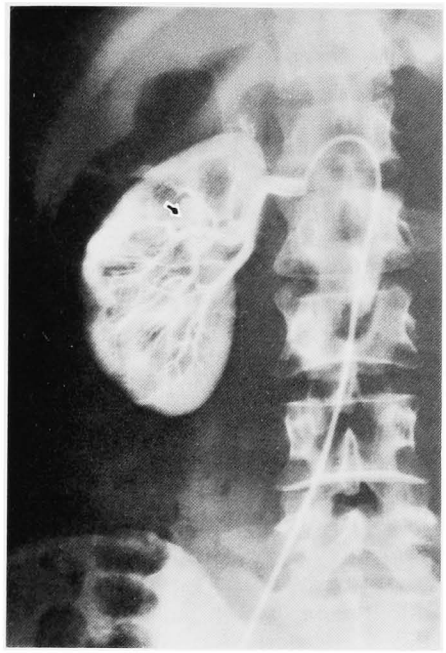

Fig. 9. Case 3-Right selective renal angiography discloses a coil made up of densely arranged and tortuous vessels (arrow)

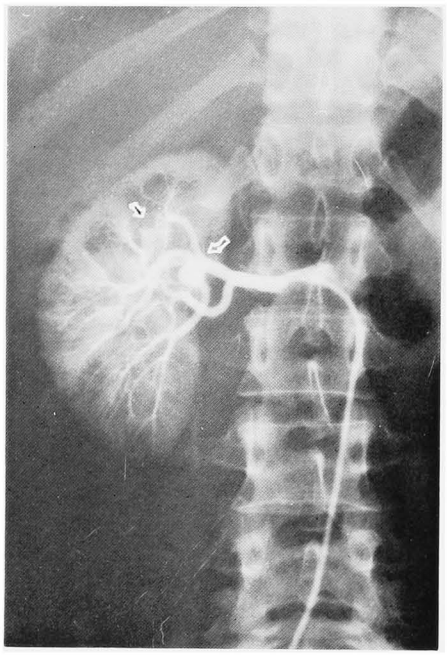

Fig. 10. Case 4-Right selective renal angiography illustrates a saccular aneurysm (9×8 mm) (white arrow) and a plexiform blush (black arrow) at the anterior upper segment

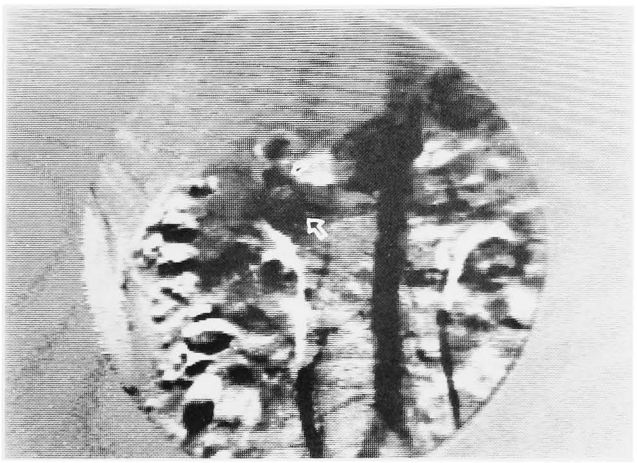

Fig. 11. Case 4—Digital subtaction angiography 12 months later suggestes no progression of the saccular aneurysm (white arrow) or hemangioma (black arrow)

resolved 3 days following the admission without any treatment, and then subsequent DIP demonstrated good visualization of the right kidney. Renal sonography and computed tomography including dynamic studies were interpreted as normal. Although RI-angiography was also within normal limits, selective angiogram of right kidney revealed a saccular aneurysm (Fig. 10). Moreover, at the upper segment, a coil of blood channels measuring 15×12 mm in size was observed 1/3 minute after injection and disappeared approximately 10 minutes later. A month later, repeated angiography of right renal artery confirmed the same findings as above, and left renal and celiac arteriography showed no abnormal findings. She has been asymptomatic with clear urine, stable blood pressure and normal plasma renin activity for 15 months. Recent digital subtraction angiography suggested no progression of the saccular aneurysm or hemangioma (Fig. 11).

DISCUSSION

Renal hemangioma is a relatively rare disease, which is considered to be a renal vascular tumor a true neoplastic growth arising from buds of endothelial cells[4] This tumor is pathologically classified with cavernous, plexiform and capillary type. It might have multiplicity with 12% and even bilateral occurrence[5]. Most such lesions are small, measuring 1 or 2 cm in diameter[6]. Nearly all patients with renal hemangioma complained of gross hematuria. The previous report in the English literature pointed out that the onset of symptoms occurred between 20 and 40 years of age in 70%[7]. There is, however, no significant correlation between age and the occurrence of this disease in the Japanese cases. Renal hemorrhage is occasionally so severe that patients might suffer from renal colic due to the passage of clots. Some cases were reported to result in vesical tamponade or anemia. More characteristically, hemorrhage from hemangioma is intermittent over a period of many years. As the site of symptomatic hemangioma is commonly pericaliceal or intrapapillary, such condition would produce papillary necrosis[8]

Renal hemangioma is usually difficult to diagnose and it may easily be missed in clinical explorations, for example on macroscopic and microscopic examinations by the pathologist[9]. Renal arteriography has been shown to be of great value for diagnosis. Tille[10] described a coil of overlapping fine vascular loops on the renal arteriograph of his case. In contrast to the irregular vessels of varying caliber in a malignant tumor, the densely collected vessels of a hemangioma are of uniform caliber and regular outline[11] Not all cases of renal hemangioma, however, demonstrate the same vascular abnormalities as above, for angiographic features could depend on the vascular component. Although renal hemangioma may be arterial and mixed veno-arterial, they are generally venous[12] When the hemangioma is more truely cavernous in type, the possibility of angiographic demonstration against the background of the normal renal parenchyma is small[13]. Augumentative techniques including a prolonged slow arterial injection, selective venography[14] or pharmacoangiography[15] might be indicative. Such lesions could occasionally disclose hypovascularity and over-all diminished contrast medium density compared to the surrounding normal parenchyma illustrated by Stanley et al. [14] They presumed that thrombosed channels, noted grossly and histologically within the tumor, could account for some of this appearance.

The renal sonogram of cavernous hemangioma might establish the solid mass pattern, while not pathognomonic, and could eliminate the possibility of a fluid filled cyst or isolated hydrocalyx[14].

Computed tomograms of 2 presented cases of cavernous hemangioma demonstrated well-demarcated round and low-density masses. It is of particular interest that they were not enhanced with contrast medium. Those of the liver are usually said to show low dense masses, and to be characteristic of nodular shadows in the low dense mass after enhancement[16]. Kamei et al.[3] also reported that enhanced tomography of their case revealed a low dense area in spite of normal plain series.

We postulate that the computed tomographic appearance of renal hemangioma might be partly related to such conditions as blood flow and vascular structure. The hypovascular or normal-appearing type on arteriography could demonstrate a low-density mass on computed tomography.

Treatment for the renal hemangioma includes nephrectomy, partial nephrectomy, papillectomy and radiation[5], wheareas all cases reported so far in the Japanese literature except for a few cases have been treated by nephrectomies. If a patient is preoperatively diagnosed to have a hemangioma, conservative management of the kidney shoud be performed as much as possible. Bischoff et al.[17] described 2 cases; conserving kidney-intraarterial superselective embolization in one case and surgical clipping of the supplying arterial branch in the other. Observation is not contraindicated in the healthy patient with mild to moderate hematuria who is otherwise clinically and radiographically well or when marginal renal function or the absence of an opposite kidney dictates such a course[5] For the future, there could be an increasing number of observation with or without the conservative treatment to the kidney. But nephrectomy remains an appropriate treatment for some cases with uncontrolled and life-threatening hemorrhage, or cases in which the possibility of other malignant renal tumors can not be eliminated in the preoperative evaluation.

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和文抄録

腎　血　管　腫

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松下　巖
大垣市民病院泌尿器科（医長：磯貝和俊）
山羽　正義・磯貝　和俊

腎血管腫の4例を報告した。良性血管性腫瘍である腎血管腫は腎動脈造影においても必ずしも異常陰影を示さないので，その診断は困難である．われわれの経験した症例のうち，2例の海綿状血管腫摘出例はCTにおいて，造影剤増強効果をもたない low-density な腫瘤影を示した．しかし，動脈造影により診断のなされた2例はCTでは異常陰影は認められなかった．このように腎血管腫の動脈造影およびCTは血管腫自体の構成血管成分により種々の像を呈するものと考えられる．